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Individual quality of life in adults with congenital heart disease: a paradigm shift

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Aims During the last decade, a paradigm shift has emerged in the measurement of quality of life, from the use of standard questionnaires towards a more individualized approach. Therefore, this study examined individual quality of life in adults with congenital heart disease and explored potential differences with those reported by matched, healthy control subjects.
Methods and results We examined 579 adults with congenital heart disease. A subsample of 514 of these patients was matched for age, gender, educational level, and employment status with 446 healthy counterparts. Individual quality of life was assessed using the Schedule for the Evaluation of Individual Quality of Life-Direct Weighting (SEIQoL-DW). Twelve domains affecting patients' quality of life were identified. Family, job/education, friends, health, and leisure time were the most prominent quality of life domains. Significantly fewer patients than control subjects considered financial means and material well-being and future to be important determinants of quality of life.
Conclusion Assessment of quality of life in adults with congenital heart disease that focusses on the individual is appropriate for obtaining in-depth information on issues relevant for patients' quality of life. This represents a paradigm shift in the measurement of this concept.

Introduction

The life expectancy of patients with congenital heart disease has increased substantially over the past decades. This decrease in mortality has elicited heightened interest in quality of life issues pertaining to patients with congenital heart disease. In addition to ongoing medical problems, many of these patients continually face specific psychosocial, educational, and behavioural challenges and concerns.[1] Indeed, for many of these patients, their heart defect impacts their quality of life on a daily basis.

There are two major approaches to measuring quality of life: the 'need approach' and the 'want approach'.[2] The need approach is a mainstay of quality of life studies. According to this approach, quality of life depends on fulfilment of basic needs, such as good health, sufficient mobility, good physical performance, adequate nutrition, and favourable shelter. In this approach, quality of life is measured using standardized and pre-defined questionnaires about components or determinants of quality of life. The relative importance of each of these components is assumed to be equal for

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all respondents. Three types of measurement are often used in this respect: (i) generic instruments, which comprehensively assess quality of life in a variety of populations; (ii) disease-specific instruments, which are developed for a particular disease or health condition; and (iii) test batteries, which employ both generic and disease-specific instruments.[3]

The want approach assumes that quality of life can only be affected by factors important to an individual.[2] For example, according to the want approach, quality of life depends on lifestyle, previous experiences, ambitions, and dreams.[2] Hence, in this approach, quality of life must be measured with instruments that permit respondents to indicate and respectively rate domains that are specifically important for their quality of life (i.e. individual quality of life). During the last decade, a paradigm shift has taken place in the measurement of quality of life, from one based on the need approach to one based on the want approach. Some now argue that the want approach is the most valid way of measuring quality of life, because it explicitly includes domains that are relevant for the respondents.[4,5] This is obviously a limitation of the need approach.

Several methods have been developed for assessing quality of life that use the want approach.[6,7] One such method is the Schedule for the Evaluation of Individual Quality of Life (SEIQoL).[6] It was developed to examine quality of life from an individual's perspective by assessing issues defined by the respondent that they feel are most important for quality of life. In contrast to pre-defined questionnaires, which assess quality of life in a more functional manner, SEIQoL reflects a more holistic view, because it considers the effects of non-disease-related aspects of life.[8] The SEIQoL, therefore, provides critical information on a patient's perspective of quality of life issues. This is important for adequate patient management in integrated healthcare programmes.

Adults with congenital heart disease constitute a relatively new and growing patient population. To meet the specific needs of these patients, understanding psychosocial issues, including quality of life, is critical for developing comprehensive healthcare programmes for this group of patients. Therefore, the goal of the present study was to identify specific issues, or domains, that most importantly affect the quality of life in adults with congenital heart disease. This was achieved in part by comparing differences in individual quality of life as defined by our study group with those defined by healthy counterparts.

Methods

Study population

The present study was part of a larger research programme examining the quality of life in adults with congenital heart disease, conducted from 28 November 2000 to 27 November 2002 at the University Hospitals of Leuven in Belgium.[9] In this 2 year period, 1535 outpatient visits by 1135 patients were performed. To be included in our study, the patients must have been diagnosed with congenital heart disease, were 18 years or older, literate, and Dutch-speaking. Patients meeting the following criteria were excluded: first-time patients of our outpatient clinic, mentally retarded patients (observed or confirmed during the clinical interview), referral or follow-up patients who had undergone percutaneous closure of an atrial septal defect or a patent foramen ovale because of cryptogenic stroke, or sensory or physical limitations to participate (*Figure 1*). Since this study used self-report questionnaires, eligible patients must have been able to read and write Dutch. Hence, quality of life of patients with mental retardation or illiterate patients were not addressed in this study, because these populations require a specific methodological approach, different from the methods used in this investigation.

A total of 716 patients met the inclusion criteria and were asked to participate in this cross-sectional, comparative study. Of these patients, 66 subjects (9.2%) declined to participate, eight (1.1%) did not participate because they felt too emotionally distressed after the visit with the cardiologist, and 13 (1.8%) were excluded for practical reasons, resulting in a sample of 629 individuals (*Figure 1*). Since the reason not to take part in the study could very well be related to the patient's quality of life, the introduction of a potential selection bias cannot be excluded. Fifty patients did not provide valid answers, and were therefore excluded from the analysis (see Results).

To explore differences between individuals with and those without heart defects, a subsample of 514 adult patients with congenital heart disease were matched with 446 healthy control subjects according to age, gender, educational level, and occupational status (*m*:*n* matching). Control subjects were volunteers recruited from a range of high schools, colleges, universities, companies, and administration organizations in our geographical region. From the remaining 65 patients, 15 and eight patients were not selected for matching because they were disabled or received special education, respectively. For patients who were retired ($n = 5$), or unemployed/housewife ($n = 30$) no matching control persons could be recruited because we could not find an organization that was willing to participate. For seven patients, no control person complying with the matching criteria was available.

Variables and measurement

On the basis of a thorough conceptual foundation, we defined quality of life 'as the degree of overall life satisfaction that is positively or negatively influenced by individuals' perception of certain aspects of life important to them, including matters both related and unrelated to health'.[10] With this definition, quality of life ought to be measured in terms of life satisfaction. The SEIQoL Direct Weighting (SEIQoL-DW)[11] was used to evaluate the aspects of life that were important for individual quality of life in our subjects (i.e. determinants of quality of life). Administration of the SEIQoL-DW includes three successive stages, comprising both qualitative and quantitative assessments:

(i) Using a semi-structured interview, we asked respondents to think about their lives and designate five domains of life that they perceived as most important for their quality of life. For each domain identified, patients were also asked to elaborate more precisely about that domain, or what in particular made that domain so important to them. This elaboration allowed the investigators to gain insight into the meaning of the identified domains.

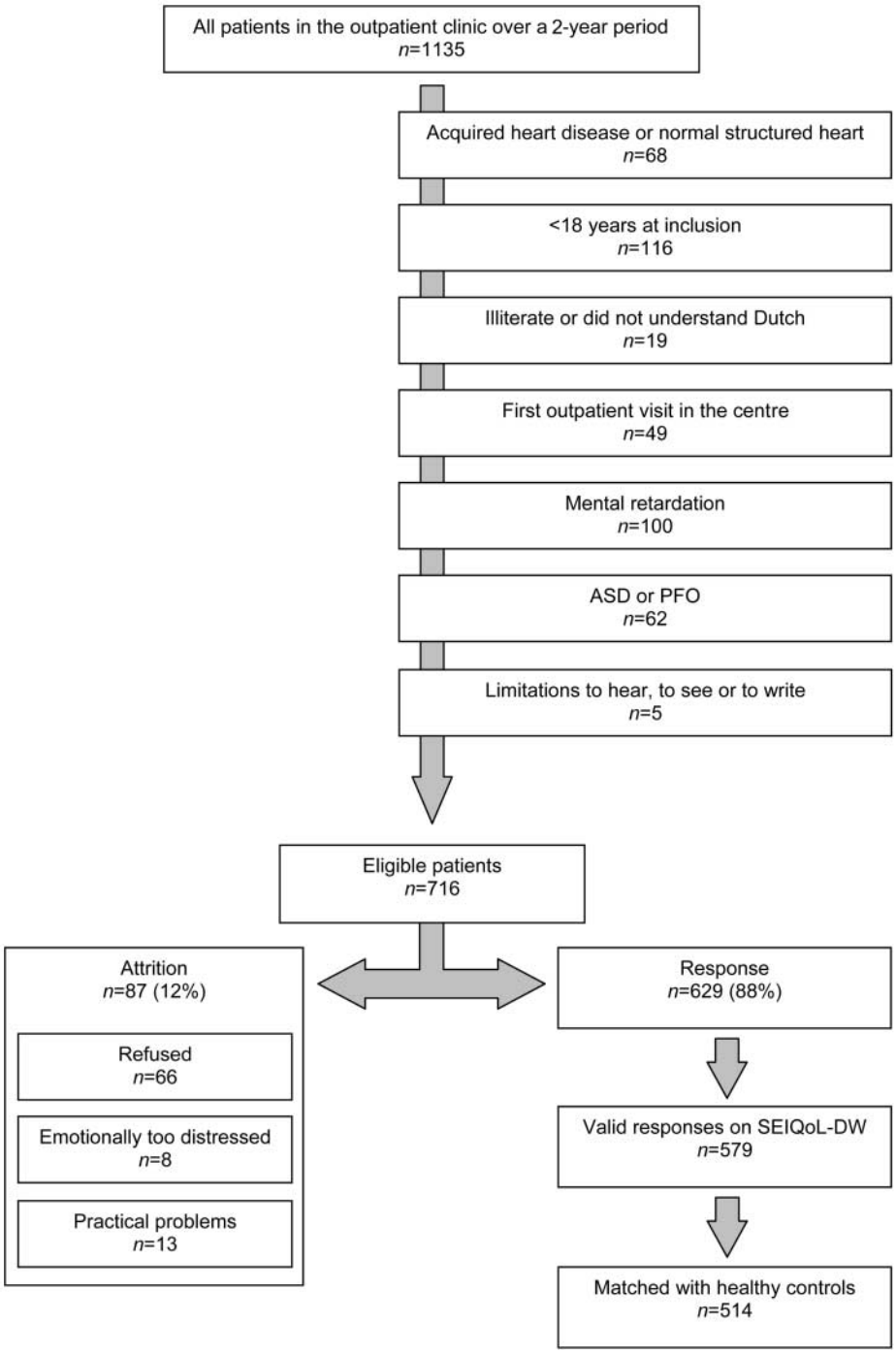

Figure 1 Flow-chart of patient selection.

(ii) Respondents were asked to rate the actual status or level of fulfillment for each specified domain by drawing a vertical box between the terms 'worst possible' (0) and 'best possible' (100). The horizontal placement of the box corresponded to the actual status or degree of fulfillment (e.g. a box drawn toward the 100 marker indicated the highest degree of fulfilment).

(iii) Finally, respondents were asked to quantify the relative importance of each domain using a coloured five-segment disc. Respondents could enlarge or reduce each segment according to their perceived relative contribution of each domain. The relative importance was expressed as a percentage.

The SEIQoL-DW permits the calculation of a single index of quality of life by summing the products of the rated level and weighting for each of the five domains. This index ranges from 0 to 100, with higher scores corresponding to higher levels of quality of life.

Although the SEIQoL-DW is known to have high face and content validity,[4,5,7,11,12] little evidence was available on other psychometric properties.[8] Using the new standards of psychometric testing, our data analyses provided additional validity evidence on test content, internal structure, and relations to other variables, as well as on the stability of the instrument.[10] Although the SEIQoL-DW cannot be considered as a measure of quality of life itself, it is a valid and reliable instrument to explore determinants for patients' quality of life.[10] Responsiveness of the SEIQoL-DW in patients with congenital heart disease might be problematic,[10] and should therefore be scrutinized in further research.

Procedure

Patients were recruited following a scheduled outpatient visit to the Adult Congenital Heart Disease Clinic. After consulting with the advanced clinical nurse practitioner and a cardiologist specialized in congenital heart defects, an independent researcher obtained informed consent from qualifying patients and provided instructions on completing the questionnaires. The researcher remained available to offer clarification if needed. Administration of the SEIQoL-DW averaged 7 min (range 3–15 min). The study protocol was approved by the local ethics committee.

Two approaches to recruit eligible control subjects were used. First, based on the matching criteria, potential control subjects were identified from the organization's personnel database. These persons were subsequently invited to participate. Second, the aim and procedure of the study was explained to the students of specific classes or to employees of the participating company by using the internal communication bulletin. Persons who were willing to participate could apply for the study. Their demographic characteristics were checked with the matching criteria. If a corresponding patient was included in the study, this control subject could participate.

Data analysis

Both qualitative and quantitative analyses were performed. Analyses were similar to content analysis techniques. For the five patient-designated domains, a standard qualitative analytical procedure was used. The domains, as well as the reasons for why they were important, were transcribed verbatim. The individual statements, in their original form, were subsequently sorted and clustered, according to common content. Each cluster was subjectively labelled according to the best description of the meaning of the statements in that cluster. For each labelled cluster, we calculated the percentage of patients mentioning the corresponding domain. To increase objectivity in the interpretation and to monitor the clustering process, peer debriefings were organized with experts in the field of quality of life and qualitative analyses. Three independent meetings with experts were arranged to reach consensus on the labelling of clusters.

Descriptive statistics of demographic and clinical variables were expressed in percentages, medians, and quartiles. Because an *m*:*n* matching was employed, conditional logistic regression on 65 strata was used if the response variable was dichotomous. Although conditional linear mixed models have been developed to be used when response variables are continuous,[13] our data did not fulfil the normality assumptions inherent to these statistics. Therefore, we calculated a mean score per group for each stratum and compared these scores between the two groups using a Wilcoxon signed-rank test. No appropriate statistics are available for the analysis of nominal level data when samples are related but unbalanced. Therefore, we used the χ^2 and Mann–Whitney *U* tests to compare some demographic and social characteristics.

All tests were two-sided. The Bonferroni correction was applied to adjust for the inflating type I error in multiple testing. For each test, 12 comparisons were made (12 domains). Therefore, the level of significance was set at $P \leq 0.004$ (0.05 divided by 12).

Since determinants of quality of life may evolve when growing older, we assessed the evolution of important quality of life domains per decade of life. For this purpose, we used the Cochran–Armitage test for trends.

Results

Patient characteristics

Of the 629 patients who were initially chosen to participate in this study, 50 were excluded because their responses were considered invalid for the following reasons: the respondents failed to completely understand the SEIQoL-DW, their answers were inaccurate, or the person accompanying the respondent interfered with the SEIQoL-DW procedure. Hence, valid data were available for 579 patients (92%), 59.9% of which were male and 40.1% were female (*Table 1*). The median age of these patients was 23 years. Given this relatively young age, the majority were unmarried and lived with their parents. Most patients were employed. Primary diagnoses exhibited most often within this sample of patients were tetralogy of Fallot, ventricular septal defect, coarctation of the aorta, aortic valve stenosis, pulmonary valve stenosis, and transposition of the great arteries (*Table 2*).

Demographic and social characteristics of the 514 patients matched with the 446 healthy control persons are compared (*Table 3*). For gender, age, employment

Table 1 Demographic and clinical characteristics of subjects with congenital heart disease

	n (%)
Gender ($n = 579$)	
Male	347 (59.9)
Female	232 (40.1)
Median age (years)	23 ($Q_1 = 20$; $Q_3 = 29$) range 18–66
Marital status ($n = 576$)	
Unmarried (living with parents)	322 (55.9)
Living alone, divorced, or widowed	56 (9.7)
Married or co-habiting	198 (34.4)
Employment status ($n = 579$)	
Student	167 (28.8)
Employed	342 (59.1)
Unemployed/looking for work	19 (3.3)
Unable to work/disability	15 (2.6)
Other	36 (6.2)
Median frequency of follow-up at Congenital Cardiology Outpatient Clinic (years)	1.5 ($Q_1 = 1.0$; $Q_3 = 3.0$) range 0.25–6

Table 2 Prevalence of primary medical diagnosis in subjects with congenital heart disease

Primary medical diagnosis ($n = 579$)	Prevalence *n* (%)
Tetralogy of Fallot	105 (18.1)
Ventricular septal defect	99 (17.1)
Coarctation of the aorta	83 (14.3)
Congenital stenosis of aortic valve	58 (10.0)
Pulmonary valve stenosis (congenital)	41 (7.1)
Transposition of great arteries (ventriculo-arterial discordance)	32 (5.5)
Combined aortic valve stenosis and aortic insufficiency	26 (4.5)
Ostium secundum atrial septal defect (ASD II)	22 (3.8)
Congenital mitral insufficiency	20 (3.5)
Univentricular heart	18 (3.1)
Double outlet right ventricle	10 (1.7)
Ebstein's anomaly	9 (1.6)
Congenitally corrected transposition of great vessels (double discordance)	8 (1.4)
Congenital insufficiency of aortic valve	8 (1.4)
Partial atrioventricular septum defect (ASD I)	8 (1.4)
Congestive cardiomyopathy/dilated cardiomyopathy	6 (1.0)
Dilatation of the sinus of Valsalva	4 (0.7)
Hypertrophic obstructive cardiomyopathy	4 (0.7)
Complete atrio-ventricular septum defect	3 (0.5)
Restrictive cardiomyopathy	2 (0.3)
Partial anomalous pulmonary venous connection	2 (0.3)
Interrupted aortic arch	2 (0.3)
Double aortic arch	2 (0.3)
Hypoplastic right ventricle	2 (0.3)
Coronary artery anomaly (ALCAPA)	1 (0.2)
Patent ductus arteriosus	1 (0.2)
Total anomalous pulmonary venous connection	1 (0.2)
Cor triatriatum	1 (0.2)
Mitral valve stenosis	1 (0.2)

status, the presence of children, the number of children, child wish, and the possession of a driver's licence, both groups were comparable [level of significance $P \leq 0.0055$ (0.05 divided by 9)]. However, significant differences were found for educational level and marital status.

Individual quality of life

The SEIQoL-DW identified 12 domains that affected patient quality of life (*Table 4*). *Family* was the most important determinant of quality of life in adults with congenital heart disease. *Job/education*, *friends*, *health*, and *leisure time* were important determinants for 48–70% of the patients. Domains such as *future*, *pets*, *environment*, and *nourishment* were important for <10% of the patients sampled.

Although some domains were important for only a few patients, the actual status or level of fulfilment of all domains was rated highly (median ≥ 75), except for *future*. Please note that, responding to the actual status of the domain *future*, respondents refer to the likelihood that this domain will be fulfilled. We observed individual variability in the actual status of some domains, as illustrated by the large interquartile range of the domains *future* (=42) and *environment* (=37).

Regarding the relative importance of the respective domains, *family* proved to be the most significant determinant of quality of life in adults with congenital heart disease, followed by *health*, *friends*, and *future* (≥20%). *Environment*, *financial means and material well-being*, and *nourishment* were less important.

On the 0–100 scale, the overall SEIQoL-DW index score for this sample of patients was 79 (Q1 = 70; Q3 = 87), suggesting that adult patients with congenital heart disease have a relatively good quality of life.

Important quality of life domains per decade of life

We found significant differences among the respective decades of life for three domains: *health*, *family* and *friends* (*Figure 2*). These data show that *health* and *family* become more important with increasing age. On the other hand, *friends* are less frequently reported by older patients as an important quality of life domain. The observed differences in the actual status and relative importance were not statistically significant.

Comparison with healthy controls

Few significant differences were found in domains important for the quality of life in adults with congenital heart disease and those identified by healthy control subjects (*Table 5*). Significantly fewer patients than

Table 3 Comparison of demographic and social characteristics of patients and healthy control subjects

	Patients ($n = 514$)	Controls ($n = 446$)	*P*-value
Gender			1.0[a]
Male	319 (62.1%)	240 (53.8%)	
Female	195 (37.9%)	206 (46.2%)	
Median age (years)	23 ($Q_1 = 20$; $Q_3 = 28$)	24 ($Q_1 = 20$; $Q_3 = 31$)	0.017[c]
Educational level			**0.003**[b]
Vocational high school	169 (32.9%)	101 (22.7%)	
Technical high school	64 (12.5%)	75 (16.9%)	
High school	45 (8.8%)	35 (7.9%)	
College	164 (32.0%)	174 (39.2%)	
University	71 (13.8%)	59 (13.3%)	
Employment status			0.959[a]
Student	165 (32.1%)	157 (35.2%)	
Employed	349 (67.9%)	289 (64.8%)	
Marital status			**0.005**[b]
Unmarried (living with parents)	300 (58.7%)	215 (48.3%)	
Living alone, divorced, or widowed	48 (9.4)	55 (12.4%)	
Married or co-habiting	163 (31.9%)	175 (39.3%)	
Children			0.089[a]
No	442 (86.2%)	334 (75.2%)	
Yes	71 (13.8%)	110 (24.8%)	
If yes, median number of children	2 ($Q_1 = 1$; $Q_3 = 2$)	2 ($Q_1 = 1$; $Q_3 = 2$)	0.448[c]
Do you wish (more) children?			0.042[b]
No	93 (18.3%)	91 (21.3%)	
Yes	295 (58.1%)	263 (61.6%)	
Don't know	120 (23.6%)	73 (17.1%)	
Driving licence			0.922[b]
No	113 (22.2%)	100 (22.4%)	
Yes	397 (77.8%)	346 (77.6%)	

[a]Conditional logistic regression.
[b]χ^2.
[c]Mann–Whitney *U* test.

Table 4 Important quality of life domains reported by subjects with congenital heart disease

	Patients choosing domain ($n = 579$), *n* (%)	Median actual status (Q1–Q3)	Relative importance, % (Q1–Q3)
Family	464 (80.1)	87 (75–94)	25 (21–30)
Job/education	403 (69.6)	75 (60–87)	16 (12–22)
Friends	346 (59.8)	82 (71–91)	21 (16–25)
Health	347 (59.1)	79 (65–90)	22 (17–28)
Leisure time	279 (48.2)	77 (60–88)	16 (12–20)
Personal characteristics and self-fulfilment	170 (29.4)	78 (64–89)	18 (15–24)
Financial means and material well-being	139 (24.0)	77 (60–88)	13 (10–19)
Important values	59 (10.2)	77 (58–86)	18 (11–24)
Future	57 (9.8)	69 (50–92)	20 (12–27)
Pets	44 (7.6)	92 (76–99)	17 (14–24)
Environment	32 (5.5)	80 (51–89)	15 (11–20)
Nourishment	23 (4.0)	93 (69–99)	11 (8–18)

control subjects considered the domains *financial means and material well-being*, and *future* to be important determinants of quality of life. Although a few control subjects considered the domains *being bereaved/loss of significant others*, *mental capabilities*, and *physical appearance/personal hygiene* to be important indicators of quality of life, interestingly, patients did not consider these domains to be important for their quality of life.

In addition to the lower proportion of patients indicating *financial means and material well-being* to be important for their quality of life, the relative importance of these domains was also significantly lower in patients than in

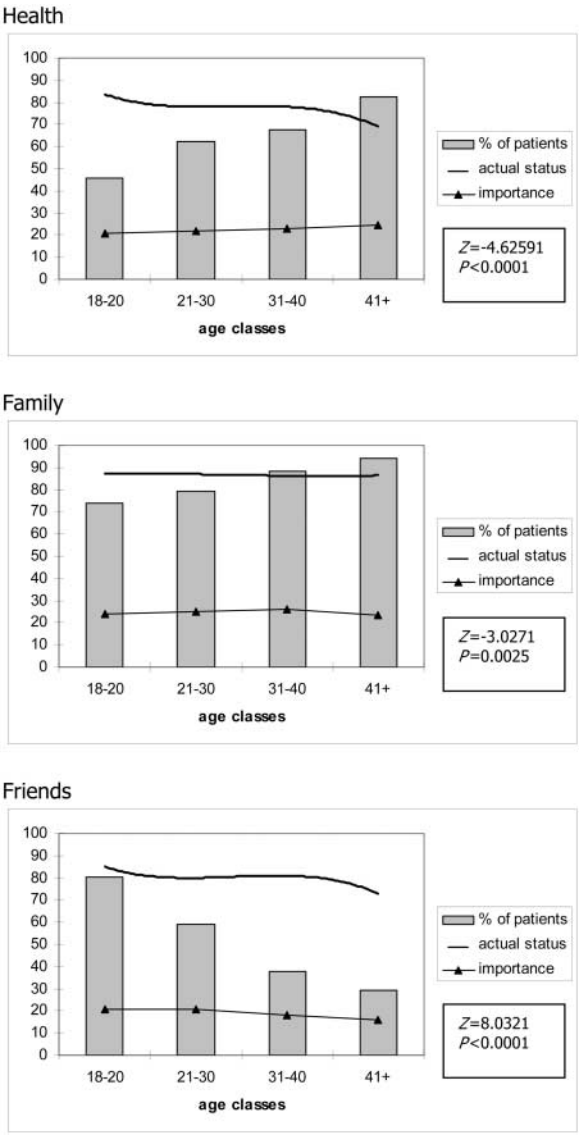

Figure 2 Important quality of life domains per decade of life.

control subjects. For the domain *future*, the relative importance was equal for both groups (*Table 5*).

Relationship to type of heart defect

We examined whether the type of congenital heart defect affected the patient's designation of domains deemed to be important for their quality of life. For this analysis, only heart defects that occurred in >5% of the sample were considered, in order to have subgroups of >30 patients. Among patients with tetralogy of Fallot, ventricular septal defect, coarctation of the aorta, congenital stenosis of the aortic valve, pulmonary valve stenosis, or transposition of the great arteries, no differences were found in the percentage of patients who identified respective domains, the actual status or relative importance. This indicates that the type of heart defect does not impact individual quality of life.

Discussion

Since mortality and morbidity of patients with congenital heart disease have decreased substantially in recent decades, interest has increased greatly in issues relating to quality of life in this patient population. To date, all published studies examining quality of life issues in adults with congenital heart disease have used the need approach.[14–32] In these studies, quality of life was measured from a functional or socioeconomic, rather than an individual, perspective. The former approach typically uses standardized questionnaires or classification systems. By assessing individual quality of life, on the other hand, researchers can take into account the unique perspective of individual patients. Evaluation of individual quality of life with the want approach is therefore more suitable because it circumvents problems inherent to standardized instruments (e.g. ratings based on arbitrary topics and equal weightings of questionnaires). Indeed, by using the want approach, a researcher recognizes that, in different individuals, different variables influence quality of life and that these variables do not equally affect all individuals.[4] Acknowledging the validity of this paradigm shift, we investigated issues important for quality of life in a large sample of adults with congenital heart disease.

We found 12 important domains that contribute to the quality of life of patients with congenital heart disease. The majority of patients identified general issues, such as *family*, *job/education*, *friends*, and *health* to be important determinants of quality of life. The relative importance was highest for *family*, then *health*, *friends*, and *future*. This indicates that while *job/education* was, for many patients, an important determinant of quality of life, it was considered less important when compared with the other nominated domains. Only a few respondents identified concerns about their *future* as an important indicator of quality of life, although its relative importance was higher compared with that of many other domains.

Quality of life research often focusses on health-related quality of life. Indeed, patients' health is consistently identified as an important determinant of quality of life. However, in the present study, only 59% of the patients with congenital heart disease identified *health* as an important determinant of quality of life. Moreover, fairly equal numbers of patients and healthy control subjects identified *health* as important. This confirms that, by focusing on health-related quality of life, investigators substantially overestimate the impact of health-related factors and seriously undervalue the effect of non-medical phenomena.[5]

Our findings identified only two significant differences in important determinants of quality of life between patients with congenital heart disease and healthy persons without heart defects. *Financial means and material well-being* was an important determinant in twice as many control subjects as patients. The relative importance of this domain was also higher in controls than in patients. Severe heart conditions or previous

Table 5 Comparison of important domains contributing to quality of life in subjects with congenital heart disease and healthy control subjects

	Number (%) of patients choosing domain			Median actual status			Relative importance (%)		
	Patients ($n = 514$)	Controls ($n = 446$)	P-value[a]	Patients ($n = 514$)	Controls ($n = 446$)	P-value[b]	Patients ($n = 514$)	Controls ($n = 446$)	P-value[b]
Family	406 (79.0)	352 (79.1)	0.882	86	82	0.091	25	24	0.006
Job/education	377 (73.3)	279 (62.7)	0.031	76	75	0.470	16	17	0.186
Friends	318 (61.9)	227 (51.0)	0.159	82	80	0.966	21	22	0.291
Health	302 (58.8)	240 (53.9)	0.161	80	83	0.038	22	25	0.014
Leisure time	250 (48.6)	172 (38.7)	0.101	77	73	0.916	16	17	0.230
Personal characteristics and self-fulfilment	156 (30.4)	135 (30.3)	0.823	79	72	0.394	18	17	0.258
Financial means and material well-being	121 (23.5)	221 (49.7)	**< 0.001**	79	75	0.086	13	15	**0.002**
Important values	54 (10.5)	77 (17.3)	0.014	80	61	0.279	18	16	0.171
Future	49 (9.5)	74 (16.6)	**0.003**	66	56	0.036	19	17	0.299
Pets	37 (7.2)	11 (2.5)	0.006	91	93	0.116	16	17	0.833
Environment	29 (5.6)	32 (7.2)	0.268	80	62	0.114	15	17	0.959
Nourishment	21 (4.1)	25 (5.6)	0.438	83	85	0.463	11	15	0.528
Bereaved/loss of significant others	0 (0)	6 (1.3)	NA		35	NA		10	NA
Mental capabilities	0 (0)	5 (1.1)	NA		64	NA		11	NA
Physical appearance/ personal hygiene	0 (0)	5 (1.1)	NA		65	NA		9%	NA

NA, not applicable.
[a]Conditional logistic regression.
[b]Wilcoxon signed rank test.

operations prompt patients to put material aspects in proper perspective. This could potentially explain why fewer patients consider *financial means and material well-being* to be important.

The present study also revealed that the type of heart defect does not impact factors deemed important for quality of life in adults with congenital heart disease, neither the actual status nor relative importance of the respective domains. This is in line with the finding that the quality of life in our patient sample was only marginally associated with the severity of the heart defect.[33] It can therefore be assumed that patients with severe conditions do not necessarily consider other aspects of life as significant.

Implications

This investigation provides crucial information for healthcare professionals to understand better the consequences of heart defects on patients' quality of life. This study goes beyond the traditional focus of functionality problems by providing a holistic outlook on living with congenital heart disease. This holistic approach is key in comprehensive, interdisciplinary healthcare programmes for these patients.

The employment of the SEIQoL-DW in this study supports the utility of this instrument to practitioners interested in measuring their patients' quality of life for clinical purposes. The time needed for completing the SEIQoL-DW is acceptable (average 7 min), and analysis—which is complex if used in research—is not necessary because only the individual responses are relevant.

Methodological issues

This article reports the application of a relatively new method of measuring quality of life in a sample of adults with congenital heart disease. The use of a patient-centred, individualized measure, such as the SEIQoL-DW, is more appropriate than standardized instruments,[34] because it provides a detailed picture of quality of life issues relevant to patients. Moreover, the SEIQoL-DW counters common problems inherent to most quality of life measures (e.g. focussing primarily on limitations and impediments, without considering positive elements that contribute favourably to quality of life). Indeed, quality of life is increasingly considered to be a positivistic concept.[10]

Despite these positive aspects, there are specific problems with individualized measures of quality of life. Some patients may have difficulty understanding the system.[8,34] In the present study, this problem was observed in 8% or 50 of the 629 patients. These patients were excluded from the analysis without affecting the sample size. Furthermore, the interpretation and analysis of data stemming from individualized measures is complex,[34] mainly because the data are qualitative. To address this issue, we used a qualitative analysis procedure that is similar to content analysis techniques. To mitigate subjectivity in the interpretation of data, final labelling was done in consensus meetings with experts in quality of life and qualitative research. Although the data collection and analysis may be complex, we have demonstrated that it is feasible to use the SEIQoL-DW in large samples. Indeed, this is the first large-scale study using an individualized quality of life instrument.

This study favourably used a large sample size. Since we enrolled patients from an outpatient clinic of a tertiary care centre, it may be argued that the sample was not representative of the entire population of adults with congenital heart disease. Many patients who are born with a rather benign cardiac anomaly are treated in the first years of life and do not need continuing check-ups at a university hospital. In addition, the strict inclusion and exclusion criteria implied that the sample was not even, as such, representative of all patients attending the outpatient clinic. We did not include patients under 18 years of age, and patients with mental retardation. The former group was excluded because questionnaires developed for adult populations are not valid to be used in adolescents. The latter group was debarred from inclusion because self-report by questionnaires requires intact intellectual abilities.

Twelve per cent of the eligible patients did not participate in the study. Since the reason not to take part in the study could very well be related to the patient's quality of life, the introduction of a potential selection bias—although limited—could not be excluded. A possible under-representation of specific heart defects in our patient sample, however, failed to affect our results since we showed that both quality of life[33] and its determinants (present study) are not influenced by the type of heart defect exhibited by the patients studied.

Although this study meets the new emerging standards of quality of life measurement, it does not fully dismiss the utility of generic and disease-specific instruments in measuring specific components of quality of life. While these instruments may not measure all aspects of quality of life, they may be useful in measuring disability related to specific diseases and effectiveness of treatment. Hence, such instruments may identify actionable items with respect to self-perceived health status of functional abilities.

Conclusion

During the past decade, a paradigm shift has occurred in the measurement of quality of life, from one based on the need approach to one based on the want approach. Individual quality of life assessment in adults with congenital heart disease provides a detailed picture of issues relevant for patients' quality of life. Although some domains were reported by only a few patients, these had high fulfilment and relative importance for them. Comparison of quality of life measures derived from healthy control subjects with those from patients with congenital heart disease indicated that both groups basically perceive the same issues to be important. This investigation provides crucial information for healthcare professionals to understand the consequences of heart defects on patients' quality of life better. Issues that arose in this study should be addressed in comprehensive healthcare programmes that aim to improve patients' quality of life.

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